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Case Report

Idiopathic spinal epidural hematoma in an infant: A case report with peculiar manifestation

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ABSTRACT

Idiopathic spinal epidural hematoma (SEH) might be an extremely rare cause of spine compression. It is most common between the fourth and fifth decades of life. It is sporadic in children and is often complicated due to the limitations of a neurologic examination and nonspecific symptoms within the pediatric population. It is typically an isolated event. Only a few cases of relapsing paraparesis due to idiopathic SEH have been mentioned in the literature in adults. We are presenting a rare case of idiopathic SEH in a 1-year-old girl whose diagnosis was deferred after a recent similar history of paraparesis occurred after her scheduled immunization and went on for around 2 weeks, after which the patient recovered completely. However, 2 months later, the patient presented again with paraparesis, and the magnetic resonance imaging revealed an idiopathic SEH. We are detailing this case to raise the plausibility of a remission and relapse phenomenon within the idiopathic SEH presentation that may mimic some neurological disorders of infancy and childhood. The present case was managed surgically and yielded full recovery.

Keywords: Hematoma, Epidural, Spine, Cord compression, Infant

INTRODUCTION

Accumulating blood within the spinal epidural space is referred to as a spinal epidural hematoma (SEH). In the absence of adequate trauma or iatrogenic injuries, SEH is categorized as idiopathic or spontaneous SEH (SSEH). Minor injury, coughing, defecation, or a Valsalva maneuver for a long time can all be connected to SSEH,[1] resulting in a tear/burst of the internal vertebral venous plexus (intraspinal veins). Alternately, it can be related to blood thinner medications or non-drug-induced coagulopathies,[2] epidural tumors, pregnancy, and underlying spinal vascular malformations.[3] However, initial SSEH presentations are often variable and atypical depending on the level, location, and size. Relapsing paraplegia with complete recovery is a rare presentation of SSEH, particularly in the pediatric population.

As mentioned by Sheng *et al.*, SEH happens due to a known cause but rarely can occur without a known one (idiopathic). They also mentioned that the prevalence of SSEH is around 0.1/100,000.[4] Alexiadou-Rudolf *et al.*, in their review of five patients with SSEH, are all aged over fifty till the age of 87.[5] Hajhouji *et al.* recently reported one case of SSEH in a toddler,

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who was a 20-month-old boy with irreducible torticollis that improved partially postoperatively.[6] Another recent case by Alkhuraiji *et al*. reported a 12-year-old child with SSEH. In addition, only 29 pediatric cases of SSEH have been reported in the literature.[7]

Relapsing paraplegia is an infrequent initial presentation of SSEH, particularly in the pediatric population. In the last decades, complete recovery from SSEH has only been reported in a few adult patients.[8]

We are detailing this case to raise the plausibility of a remission and relapse phenomenon within the idiopathic SEH presentation that may mimic some neurological disorders of infancy and childhood. It is important to pick up this tricky disease entity in this particular age group as it can easily be missed and permanently damage the spinal cord affecting the entire patient's life.

CASE REPORT

A 12-month-old girl, a product of a normal vaginal delivery, with no significant past medical and surgical history, presented with a history of inability to move her lower limbs, crying on handling that advanced quickly during a couple of days before presentation. Two months before her presentation, the patient developed similar symptoms of not moving her lower limbs after receiving a routine vaccination. The symptoms continued for about 14 days, after which the patient resumed normal walking. The parents denied any significant injury or trauma history and noted that she was physically well and active in the interceding periods. There was no individual or family history of bleeding issues, blood thinners treatment, or intrusive spinal procedure history.

On admission, the patient was fully awake, alert, and afebrile. However, she was irritable and cries with handling. Pain sensation appeared to be diminished in both lower limbs, and her deep tendon reflexes were brisk. The neurological examination uncovered a symmetrical weakness in the lower limbs with a maximum power of 2/5. Regarding upper limbs examination was normal and moving spontaneously. All her blood tests, including her coagulation profile, were normal. From history, examination and laboratories, iatrogenic, traumatic, and infectious causes are preliminary excluded but other causes such as epidural tumors, acute herniated intervertebral disc, arteriovenous (AV) malformation, and SSEH needed further investigations. Magnetic resonance imaging (MRI) of the brain and whole spine was carried out on day 2 of admission. The MRI findings were normal for the brain but demonstrated an epidural hematoma involving the posterior and right anterolateral aspects of the spinal canal extending from the level of T1–T3, with an extension to the right T2–T3 neural foramina. This was causing severe compression on the spinal cord with a left anterolateral displacement of the cord. The hematoma measured 2.1 × 1.4 × 2.5 cm in the maximum transverse, anteroposterior, and craniocaudal dimensions, respectively. A mixed signal intensity with predominant hyperintensity was seen in the T1-weighted images, and a low signal was seen in the T2-weighted images. There was no evidence of contrast enhancement within the lesion, and no other lesions were identified [Figure 1].

The patient was seen, investigated, and followed by the pediatric neurology team. After ruling out other possible neurological disorders, the patient was referred to the spine surgery team after finding the hematoma in the MRI and urgent surgical decompression with one-level laminectomy at the T2 level and evacuation of the hematoma was performed. The patient promptly began to move her legs postoperatively. An etiologic workup was performed with the assistance of pediatric hematology, and no underlying disease could be identified. Histopathology of the excised specimen confirmed the diagnosis of hematoma.

The patient was discharged home after 3 days. Three weeks later, she presented to the clinic walking freely and could sit and stand independently. One year later, she exhibited

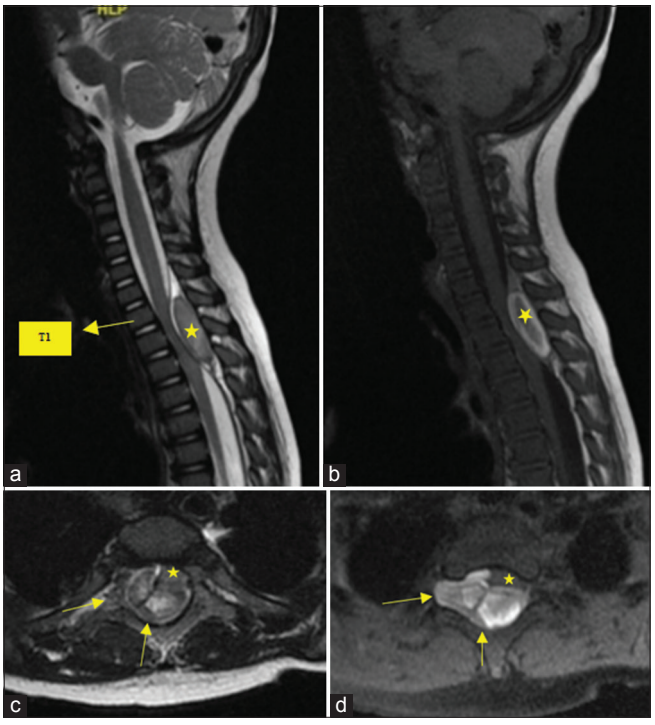

Figure 1: (a and b) are sagittal T2, T1 Magnetic resonance imaging (MRI) images showing upper thoracic spinal epidural hematoma (SEH) (star) extending from T1 level (arrow) to T3 with significant spinal cord compression. (c and d) are axial T1, T2 MRI showing SEH involving the posterior and right anterolateral aspects of the spinal canal at T2 level with extension of the hematoma to the right T2–T3 neural foramina (arrows). The spinal cord compressed with a left anterolateral displacement of the cord (star).

complete neurological recovery at her visit to the clinic with no relapse following the next dose of vaccines.

DISCUSSION

Vaccines could disturb blood homeostasis as a side effect. Although it is rare, some vaccines could cause low platelets. Measles-mumps-rubella (MMR) vaccine is rarely associated with immune thrombocytopenic purpura (ITP), it is self-limited, non-life threatening, and susceptible children with ITP should be immunized with MMR at the recommended ages.[9] This is not the cause in our case.

Our case had an infrequent presentation with an episode of paraparesis to which her primary care physician did not attach great importance due to her recent vaccination history. Then, the patient recovered completely before the second episode of paraparesis 2 months later when she was diagnosed with SSEH, once again, with some delay, because her presentation of remission and relapse was thought to be due to a neurological disorder. The patient's initial presentation was not investigated with MRI, so we cannot explain the relapsing paraparesis of her complete spontaneous recovery. At the beginning stage of SSEH, we believe the hematoma might have placed less pressure on the spinal cord. It may have spread along the spinal epidural space that contains fat and areolar tissue, which may have resulted in the successive decompression of internal pressure.

It is crucial to recognize the location of the hematoma compressing the cord for decompression. It is worth mentioning that the posterior epidural space is affected more than the anterior space due to different ideologies.[10-12] Unfortunately, our infant here had compression on the cord from the posterior and anterolateral (combined) aspects, which is once again a peculiar manifestation that needs to be taken care of.

In agreement with our treatment plan, which was single-level (T2) laminectomy at the area of compression that resulted in a good outcome of complete resolution of symptoms, Rajz *et al.* did a retrospective review of two tertiary academic centers for a total of 17 patients of different ages with SSEH utilizing the same treatment option, which is laminectomy but with variable outcomes ranging from complete resolution to quadriplegia.[13] One of the cases reported by Alexiadou-Rudolf *et al.* is an 87-year-old female who had SSEH at T11–12 secondary to AV malformation. The patient was managed conservatively, but unfortunately, the patient did not improve.[5]

CONCLUSION

Our case highlights a tall plausibility of a remission and relapse phenomenon within the presentation of SSEH in this infant patient with an odd, combined location. It is crucial to do a careful clinical evaluation and neuroimaging tests to get a diagnosis followed by prompt intervention to avoid permanent neurological compromise.

ETHICAL APPROVAL

Approved by Research Ethics Committee at Prince Sultan Military Medical City, Riyadh, Saudi Arabia. IRB number HP-01-R-079, dated on March 22, 2021.

AUTHORS' CONTRIBUTIONS

AA and IO share in the diagnosis and the management of this reported patient, AA analyzed and interpreted the patient data regarding the clinical presentation. IO performed a literature review for similar cases, IO was a major contributor to writing the manuscript. KA helped in the literature review discussion and contributed to writing the manuscript. All authors have critically reviewed and approved the final draft and are responsible for the manuscript's content and similarity index.

USE OF ARTIFICIAL INTELLIGENCE (AI)-ASSISTED TECHNOLOGY FOR MANUSCRIPT PREPARATION

The authors confirm that there was no use of Artificial Intelligence (AI)-Assisted Technology for assisting in the writing or editing of the manuscript and no images were manipulated using the AI.

DECLARATION OF PATIENT CONSENT

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parent has given his consent for the patient's images and other clinical information to be reported in the journal. The parent understands that the patient's name and initials will not be published, and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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CONFLICTS OF INTEREST

There are no conflicting relationships or activities.

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